

## Chronic otorrhea with otalgia and acoustic hypesthesia in a Romanian patient

Maria Angeles Asencio\*; Maria Huertas; Jorge Gaitán; Rafael Carranza; José Maria Olalla; German Lopez-Larramona

**\*Corresponding Author: Maria Angeles Asencio**

Microbiology Department, La Mancha Centro General Hospital, Avenida de la Constitución 3, 13600 Alcázar de San Juan, Ciudad Real, Spain.

Phone: +34-926-580567, Fax: +34-926-546882; Email: +34-926-546882

### Abstract

Tuberculous otitis media is exceptional, with several factors that converge in the difficulty of its diagnosis. Among them, the low index of suspicion, the variability of the symptoms, the low incidence of association with respiratory affection in immunocompetent patients and the false negatives of the culture. Therefore, the diagnosis and appropriate treatment can delay for months or even years. It is often followed by increased risk of serious intracranial complications.

We report the case of a Romanian patient with an 8-year history of tuberculous otomastoiditis who presents cholesteatoma, moderate deafness and a persistent opening of a postauricular fistula.

### Keywords

Chronic otorrhea; tuberculous otitis media; otomastoiditis; chronic otitis media.

### Introduction

The Chronic Otitis Media (COM) is an inflammatory process of the middle ear's mucoperiosteal lining and the mastoid, with an insidious onset, slow course and lasting for at least 3 months, which more frequently affects children under the age of 15 [1]. The most characteristic symptom of COM is the discharge, persistent or recurrent, that may be associated to eardrum perforation, acoustic hypesthesia, vertigo and tinnitus, as well as the possible existence of cholesteatoma. It is usually a mixed polymicrobial infection with anaerobic (*Bacteroides*, *Fusobacterium*, *Prevotella*, *Porphyromonas*) and aerobic flora (*Staphylococcus*

*aureus*, *Pseudomonas aeruginosa*, *Proteus sp.* y *Escherichia coli*) [2].

The tuberculous otitis media is a rare form of presentation in developed countries, representing less than 1% of COM infectious cases [3]. The infection is characterized by profuse and painless otorrhea, numerous eardrum perforations and facial paralysis (classic triad), with occurrence of pale granulation tissue and bone necrosis; however, there are few patients that present the classic symptomatology [3-7]. This low specificity and heterogeneity of the symptoms along with the poor suspicion results in delays of the diagnosis of even several years [3-5,7,8], increasing the probability of serious events as facial paralysis (more frequent in children), hydrocephalus, brain abscess, tuberculomas or meningitis [5-8].

## Case presentation

A 51-year-old Romanian male, that has been living 12 years in Spain, with intermittent otorrhea and an 8-year history of right ear moderate conductive acoustic hypesthesia. An atelectatic COM was diagnosed and subsequent bone destruction with solution of continuity and mastoid opening contact in 2014. The performed scintigraphic study was compatible with right mastoids. Apart from being a 20 daily cigarettes exsmoker for 5 years, he does not present any other relevant clinical history.

After two tympanoplasties and several cycles of topical corticoids, antibiotics and empirical antifungals, the patient presents to ER with acute intense otalgia with abundant purulent discharge, which prevents to see the eardrum, and acoustic hypesthesia. The treatment is prescribed with prednisone 60 mg IM, oral amoxicillin-clavulanic 875/125 mg and oral ibuprofen 600 mg every 8 hours, and the patient is referred to ENT.

The petrous portion of temporal bone CT showed an occupation of the right middle ear and mastoid cells with destruction of the scutum, back wall and possible erosion of the right back semicircular canal and tympanic membrane utricle compatible with cholesteatoma (Figure 1). A month later, a right mastoidectomy is performed obtaining tissue samples that are sent to the laboratory in order to carry out a microbiological and histological study. An increase of the discharge is observed with an opening of a recurrent postauricular fistula.

In the Gram staining, microorganisms were not observed and both the aerobic and fungi cultures were negatives. In the auramine staining, a great number of acid-alcohol-fast bacilli were observed, so a real time PCR was carried out over direct sampling, amplifying *Mycobacterium Tuberculosis Complex* (MT), without preliminary screening of rifampicin resistance. The Mantoux screening test was positive (20 mm) and the patient does not report neither personal nor family history of Tuberculosis (TB).

The histological study shows a large number of granulomas with caseous necrosis (Figure 2) and Langhans cells consistent with tuberculous etiology. After 7 days, growth of MTBC was obtained, which was susceptible to all first-line tuberculostatic agents. A chronic tuberculous otomastoiditis is diagnosed, consequently a serial of sputum for the study of mycobacteria (which tested negatives), a chest X-ray (in which it was not objectified cavitated lesions), and a HIV test (that resulted negative), were requested. The chest

X-ray showed paraseptal emphysema with predominance over the upper lobes and a node of 23 x 18 mm on the right vertex of a possible granulomatous origin. There are no detectable mediastinal or hilar nodes nor axillary node growths of significant size. It is initiated a treatment with isoniazid, rifampicin, pyrazinamide and ethambutol during the first two months and then isoniazid and rifampicin until 12 months are completed, with a good tolerance and a positive development. The patient is clinically stable, with no mastoid discharge nor alterations in the renal function and hepatic profile.

## Discussion

Around 20% of the TB cases are extrapulmonary [9]. The affectionation of the ENT territory has as most frequent manifestation the cervical lymphadenitis (95%); in a much lesser extent *M. tuberculosis* can be located in larynx, pharynx or cavum, ear, nasal and oral cavities [10]. In our hospital, the 3.5% of the TB are located in the ENT territory (5 ganglionic and one COM), which entails a 24% of the extrapulmonary TB; in our area, these correspond to 15% of the total in the last 13 years.

The transmission mechanisms of the tuberculous otitis media include the hematogenous spread from another focus, the ascendant ductal tract from the Eustachian tube or the direct introduction from the external auditory canal through an eardrum perforation [11], although it has been even described the congenital infection, whether from the placenta or during the pass through the birth canal [12]. However, the most common production mechanism is through secretions towed by the cough, because it can coexist with an active pulmonary TB and cervical adenopathies. Due to the conventional culture of the discharges is often negative, a tissue sample for the study of mycobacteria and histologic examination must be taken. In this case, the delay of suspecting a tuberculous etiology was of 8 years, developing into a cholesteatomatous otomastoiditis, moderate deafness and an opening of a postauricular fistula.

The absence of respiratory symptomatology in our patient, with positive Mantoux, a serial of negatives sputum and the observation of a granulomatous pulmonary node in the right vertex, indicate a past pulmonary TB, not confirmed by the patient. The pulmonary TB is the most frequent source focus [5], although, in a 40-50% of the tuberculous otitis in immunocompetent patients it is not demonstrated the illness in other locations [6,7]; in these cases, the Eustachian tube is considered as the possible portal of entry of the bacillus.

Although the laboratories continue basing the TB diagnosis in the smear and the culture, it is essential to have a molecular biology technique that provides a reliable and fast result at short notice, because the microscopy, despite its rapidity and low cost, only detects half of the TB cases, it does not distinguish the involved species of mycobacteria and it is unable to guide which TB agent to administrate [13]. It is recommended to use molecular fast tests for the resistance studies, at least in patients with a higher risk of suffering from a resistant TB: wrong adherence to a previous TB treatment, contact with resistant TB patients and provenance of countries with a multi-drug resistant TB incidence equal or greater than 5%.

The histological study is another key element to form the puzzle and, in some cases, it is the only fact in

which the diagnosis is based [5-7]. Thus, the existence of a granulomatous tissue with epithelioid cells, giant Langhans cells, caseous and necrotic areas, together with the clinic can be enough for the diagnosis given the low microbiological performance of the extrapulmonary samples [9]. It can be related to the difficulty in obtaining the sample, the low concentration of the bacillus in the middle ear (paucibacillary lesions based on little optimal conditions for the growth), as well as the previous treatment with aural antibiotics (topic gentamicin or fradiomycin) weakly antituberculous [3], that mask the presentation of tuberculous otitis media. Within the differential diagnosis, the mycotic otitis, granulomatosis with polyangiitis, sarcoidosis, syphilis, amyloidosis and the squamous cell carcinoma must be discarded.

The prevalence of resistant MT strains is usually higher in immigrants; in our area, they mainly come from Morocco and Romania, recommending to initiate therapy with 4 antituberculous until the result of the antibiogram is obtained [14]. Nevertheless, the isolated strain showed susceptibility to all the first-line tuberculostatic agents. The duration of the treatment is variable and many times it depends on the patient's evolution, varying from 6 months [3,8] to 12 or even 18 months [5,6]. Although there is no consensus in the bibliography, the complicated cases require to combine antituberculous and surgery [5,15], which provides, moreover, adequate samples for a fast and precise diagnosis of the illness. Despite the patients often respond well to the treatment [3,5,15], in some cases irreversible sequelae may remain, being the most frequent the hearing loss [6].

## Conclusions

The tuberculous otomastoiditis is an unusual entity that is underdiagnosed, with a growing number of cases in adults [3,6,14]. The presence of a persistent otorrhea with a very torpid and refractory evolution to the usual antibiotic treatment must give rise to suspicion a tuberculous etiology, especially if the patient comes from a country with a high TB endemic or has been in contact with the illness. Therefore, it would be advisable to perform a routine Mantoux to those patients. To have a rapid diagnostic technique in the laboratories is imperative to offer a quality result.

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**Authors Information:** Maria Angeles Asencio<sup>1\*</sup>; Maria Huertas<sup>1</sup>; Jorge Gaitán<sup>1</sup>; Rafael Carranza<sup>1</sup>; José Maria Olalla<sup>2</sup>; German Lopez-Larramona<sup>3</sup>

<sup>1</sup>Microbiology Department, La Mancha Centro General Hospital, Spain.

<sup>2</sup>Pathology Department, La Mancha Centro General Hospital, Spain.

<sup>3</sup>Internal Medicine Department, La Mancha Centro General Hospital, Spain.

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